



TELEPRACTICE FOR PEDIATRIC DYSPHAGIA: A CASE STUDY

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ABSTRACT

A closed-ended intensive pediatric swallowing telepractice program was developed and piloted in one pediatric patient with Opitz BBB/G and Asperger's Syndromes, oropharyngeal dysphagia and aerophagia. The present study is a case report. Outcome variables included behavioral, swallowing and quality of life variables, and were assessed at baseline and at the end of the four-week program. Selective variables were also assessed at a follow-up family interview four weeks post program completion. Over the four-week intervention period, the patient demonstrated substantial improvements in: oral acceptance of eating-related objects and a variety of foods (behavioral variable), timing of voluntary saliva swallows and aerophagia levels (swallowing variables) and quality of life. Follow-up interview analysis showed that most skills were retained or improved one-month post intervention. This intensive telepractice program proved to be feasible and effective for this pediatric patient with dysphagia.

Keywords: Dysphagia, telemedicine, telepractice

Feeding or swallowing disorders (i.e., dysphagia) can be defined as difficulty placing, receiving, maintaining and transferring food and liquids from the mouth to the esophagus and the stomach. If left untreated, feeding and swallowing disorders may lead to serious complications, including social isolation, respiratory compromise, aspiration pneumonia, malnutrition, or even death (Kirsch & Sanders, 1988; Mion, McDowell, & Heaney, 1994). In pediatric patients typical development of feeding and swallowing skills can potentially be disrupted by a variety of disorders and conditions, including developmental disability, prematurity, cardiopulmonary, neurological and gastroesophageal disorders, anatomic malformations and/or genetic syndromes (Field, Garland, & Williams, 2003; Lefton-Greif & Arvedson, 2007). Pediatric feeding and swallowing can be affected in multiple ways, including impairments in sensorimotor function, inadequate development of the typical eating/feeding milestones and the required subskills for each milestone, and disruptions of the eating pragmatics, i.e., the social behaviors required during eating (Sheppard, 2013). Such disruptions may lead to malnutrition, growth and development failure, and poor health in children (Lefton-Greif & Arvedson, 2007).

Summaries of investigations on the prevalence of feeding and swallowing disorders in children indicate that 33% to 80% of children with developmental delays and 46% to 89% of children with Autism Spectrum Disorders experience swallowing and feeding challenges

(Burklow, Phelps, Schultz, McConnell, & Rudolph, 1998; Lefton-Greif & Arvedson, 2007; Linscheid, 2006; Nadon, Feldman, & Gisell, 2013). Main reasons for this variability in prevalence estimates include differences in assessment methods, and studies' methodologies, as well as the inherent variability of these pediatric populations. Despite this variability, taking into consideration these numbers, as well as the devastating complications of swallowing and feeding disorders in one's quality of life, and growth and health status, the need to optimally diagnose and treat them is crucial.

Since the inclusion of feeding and swallowing disorders in the Speech and Language Pathology (SLP) scope of practice, an increasing need for specialized dysphagia SLPs has been apparent. Expertise in treating feeding and swallowing disorders is an area in which adequate training requires, apart from the traditional undergraduate and graduate level coursework, extensive supervised and independent experience, continued education efforts, advanced training in multiple medical applications, for example use of videofluoroscopy and endoscopy, as well as collaboration with other medical professionals (American Speech-Language-Hearing Association, 2002). The number of adequately trained SLPs in dysphagia assessment and management, and the associated availability of services, is limited in the US (Coyle, 2012) and in other countries.

All the above create an increased need to develop comprehensive telepractice programs that will allow thorough evaluation procedures and treatment delivery paradigms for those patients with swallowing disorders who live in underserved communities or for patients who are so complex that require expert involvement that is not widely available. To date, there is a handful of research studies and one clinical report on the use of telepractice in the diagnosis and rehabilitation of swallowing disorders (Georges, Belz, & Potter, 2006; Malandraki, McCullough, He, McWeeny, & Perlman, 2011; Malandraki et al., 2013; Perlman & Witthawaskul, 2002; Sharma, Ward, Burns, Theodoros, & Russell, 2013; Ward et al., 2009; Ward, Sharma, Burns, Theodoros, & Russell, 2012). These studies present the feasibility of using telepractice in dysphagia assessment with few also reporting positive reliability results when comparing telepractice to in-person evaluation procedures (Malandraki et al., 2011, 2013; Ward et al., 2009, 2012). This line of research so far has focused on adult dysphagia populations and has used rather expensive, dedicated and custom-made telepractice systems. To our knowledge, as of today there are no studies investigating the feasibility, validity and reliability of providing dysphagia telepractice services in pediatric populations.

In the present study we report a closed-ended intensive pediatric dysphagia telepractice program developed in a dedicated Dysphagia Research Clinic. This program uses freely available but secure telepractice software and was pilot tested in one complex pediatric patient from England whose family requested our expert help. The main purpose of this case study was to examine the feasibility of providing dysphagia treatment via telepractice in this pediatric patient; secondarily, we aimed to examine whether this treatment program was effective for this child.

METHODS

PROGRAM DEVELOPMENT

The telepractice program presented herein is part of a dedicated Dysphagia Research Clinic (DRC) housed in a University setting and was initiated in January of 2013. DRC includes a dedicated Telepractice Suite that houses the relevant software and hardware systems. Technological applications consist of a Windows-based personal computer (Dell, RAM: 3.25 GB, 3.00 GHz Core Duo Processor) connected to a 37-inch monitor (Samsung TV monitor, 1080 p LCD HDTV), external speakers, a stand-alone microphone and an external web-camera (Logitech Webcam C930, HD 1080p). Internet connection is achieved via a high-speed wired connection (speed: 1Gbps).

TELEDYNAMIC SOLUTION

Our telepractice computer has three tele-applications installed, including Adobe Connect, VSee and Skype. After testing multiple applications, VSee was selected as our teledynamic solution for this program for its ease and consistency, and because it allows for the following security defenses and functional uses: (a) it uses FIPS 140-2 certified 256-bit AES end-to-end encryption (i.e., there is no in-between server where the data are stored or transferred) and only the clinician (on one end) and the patient (on the other end) have access to the videoconferencing session; (b) it does not require network administrator permissions, and thus there are fewer opportunities for administrators to listen in to the sessions; (c) it does not require the creation of a public profile by the user; (d) it has screen sharing capabilities which can be useful in evaluation and treatment protocol applications and reward systems; and (e) its basic format is available for free.

ADDITIONAL DEFENSES AND CONSENTING

Additional defenses that we have established to protect patients' privacy and confidentiality include installment of current anti-virus and anti-spyware protection, and password protection. Patients and caregivers are fully informed through the informed consent process about the exact use of the technology selected, the benefits of this type of treatment delivery via telemedicine as well as all potential risks related to their privacy and confidentiality, and about all measurements taken to decrease these risks. For this specific patient, the patient's mother communicated with the clinicians (after being referred by a local SLP) and inquired about our treatment research program. She was subsequently informed over e-mail about the consenting process and the principal investigator (first author) also talked to her over the Internet to address any questions. The consent forms were sent via e-mail to the mother, who read and signed the forms and returned them to our clinic prior to any initiation of services. On the first day of our online communication with the patient, an assent form was also read to the patient (written in simple and age-appropriate language) requesting his verbal permission to use this data for research purposes and verbal permission was granted. This research study has been approved by the Teachers College Columbia University Institutional Review Board.

TREATMENT DELIVERY PARADIGM

The treatment paradigm tested in the present study is a closed-ended intensive treatment program with 1-hour sessions completed twice a week for four weeks and daily 30-minute homework practice with family involvement. Emerging literature suggests that intensive interventions that incorporate principles of activity dependent neuroplasticity (e.g., intensity, specificity, saliency) (Kleim & Jones, 2008) and motor learning (Schmidt & Lee, 2005) are effective in the treatment of swallowing and speech disorders (McKirdy, Sheppard, Osborne, & Payne, 2008; Pitts et al., 2009; Ramig, Sapir, Fox, & Countryman, 2001; Robbins et al., 2005; Robbins et al., 2007; Sapienza, 2008) and may be more or equally effective than longer treatment paradigms (Spielman, Ramig, Mahler, Halpern, & Galvin, 2007). The closed-ended character of the program was necessitated by the University summer session calendar when our treatment clinic is in session.

CASE REPORT

HISTORY

AB (initials used are fabricated to protect patient's privacy and confidentiality) is a 6-year, 6-month old male with complex medical history, who at the time of referral had aged out of locally available services for feeding therapy. Primary diagnoses included Opitz BBB/G Syndrome and Asperger's Syndrome. Anatomical anomalies seen in AB in association with the Opitz BBB/G syndrome were Level III laryngo-esophageal cleft, subglottic stenosis and unilateral cleft of the upper lip and alveolar ridge, all of which had been surgically corrected in the first five years of life. The patient was also status post tracheostomy with tracheostomy decannulation occurring ten months prior to the initiation of the program. Recent brain MRI revealed mild deficiency/hypoplasia of the inferior cerebellar vermis and mild thinning of the splenium of the corpus callosum, but no other neurological abnormalities.

AB had been totally dependent on gastrostomy tube (GT) feeding for nutrition and hydration for four years and until two weeks prior to initiation of our program. He had partially transitioned from GT feeding to a liquid diet taken only by straw in the last two weeks. At the time of the evaluation, he fed himself this liquid diet at a rate of 32mL/minute; he was not accepting or swallowing puree or other solid foods and he was not eating from spoon or fork or drinking from an open cup. Thus, his oral feeding development was significantly delayed for his age (6 years 6 months). In addition, there were concerns with persistent aerophagia (i.e., excessive air swallowing), which became more apparent post decannulation and required frequent air suctioning from the GT to alleviate stomach distention and feelings of pain and discomfort.

He was referred to the DRC for assistance with developing eating skills and tolerances for solid foods. Although he had recurrent bouts of pneumonia in the past, he had been free from a pneumonia diagnosis for the past three months. A Videofluoroscopic Swallowing Study (VFSS) had been performed 2 months prior to initiation of the telepractice program. It was performed in his home country at the request of our team prior to his transition from GT to the liquid, oral diet. The VFSS revealed no aspiration or penetration and no apparent oral-pharyngeal swallowing difficulties on the small boluses of liquids that he was able to swallow during the examination. Medications at baseline included Azithromycin, Movicol and Ducolax. AB also has multiple food and environmental allergies. He has no apparent gross or fine motor deficiencies or difficulties; and speech and language appear to be developing normally. He is in a mainstream classroom with one-to-one support. At the initiation of this program, his only therapy service was occupational therapy in which he received "sensory integration therapy".

DESIGN

This report is a case study. A one-time baseline evaluation was performed, followed by a closed-ended, intensive, treatment program, with eight, hour-long sessions over the course of four weeks; a post-intervention evaluation at the end of the four-week program; and a follow-up family interview four weeks post program completion.

BASELINE EVALUATION

The baseline evaluation was completed online prior to the first therapy session and was performed by a trained graduate student (second author) and her clinical supervisor who is an expert in pediatric dysphagia (last author). The following parameters were assessed (Sheppard, 1994): oropharyngeal sensorimotor skills; sensory oral tolerance; integrity of voluntary cough and swallow; and feeding, swallowing and eating behaviors during feeding trials. All clinical observations were judged visually via a 4-point clinical subjective scale (normal = no deviation is noted; mild = mildly deviated from normal, but still functional; moderate = overt deviation from normal, but some function is present; severe = profound deviation from normal or complete inability to perform the task). All clinicians in our clinic use these definitions, which have been established by the first author, and reliability among the clinicians (including student clinicians) has to be high (>80% agreement) before they can perform evaluations.

Results at baseline revealed the following symptoms:

- mild left-side facial and lingual weakness as evidenced by mild left-side facial drooping and

reduced ROM on movement of the tongue to the left;

- oral defensiveness as evidenced by hesitation or refusal to place eating-related objects (e.g., lollipop, spoon, toothbrush) and solid foods in his mouth;
- voluntary cough upon command of moderate strength;
- significantly delayed initiation of voluntary saliva swallows upon command (delay at baseline: ~27 seconds); a latency measure was utilized for this task and consisted of the duration from a verbal command “swallow” to the time the swallow was initiated, as indicated by visual elevation of the hyolaryngeal complex and/or palpation of the complex by his mother (when visual inspection was not adequate). For us to be able to view AB’s swallows, we had requested that he sit on the side (so we can view him laterally). We had also requested his mother to place her fingers on the lateral surface of his neck in order to provide validation of his swallows. This was completed after training the mother on “how a swallow feels” on her neck and AB’s neck. The duration was measured and reported with a stop-watch only for the swallows for which there was adequate visualization of the thyroid notch and its movement. At times, a tape was placed over his thyroid notch to enhance visualization. Reliability measures were completed for this measurement (reported later).
- oral acceptance and consumption of two types of thin liquid (a yogurt drink and a fruit smoothie) by straw, via taking 2-3 sequential sucks prior to a swallow and a breath; and,
- refusal to accept any other consistencies and volumes of foods and liquids. According to family report, this was typical of his eating behavior.

Additionally, at baseline, AB’s mother reported that he had been consuming four types of drinks (of eight flavors) of one consistency (thin liquid) only by straw; and that he exhibited severe aerophagia (i.e., excessive air swallowing), as apparent on observation and indicated by the need to suction an average of 12 liters of air and saliva from his feeding tube daily (1 liter every hour during daytime). Suctioning was performed by the mother or the father of AB. Need to suction was evaluated by the parents and was determined by the amount of distention of AB’s stomach and his level of discomfort/pain.

OUTCOME VARIABLES AND INSTRUMENTATION

The main outcomes included behavioral variables, swallowing/eating variables and quality of life variables. These were: (a) oral acceptance/tolerance of eating-related objects and a variety of foods/flavors (behavioral

variables), (b) voluntary saliva swallows, rate of intake, and aerophagia level (swallowing/eating variables), and (c) quality of life relating specifically to eating and swallowing (quality of life variable). All variables were evaluated at baseline and post-treatment. A few were additionally obtained via self-report at the post four-week follow-up family interview.

Oral acceptance/tolerance of eating-related objects was assessed via a latency measure, including the duration of the time from a given relevant command to the placement of each object fully inside the patient’s mouth. We consider this latency measure as an indication of his hesitation level and thus an indirect measure of oral acceptance of eating-related objects. Oral acceptance/tolerance of a variety of foods/flavors was assessed via a frequency measure, including the number of different foods/flavors that AB successfully accepted and tolerated (swallowed) at baseline, post treatment and at the four-week follow-up. This frequency measure was a combination of the number of different foods accepted/tolerated in the evaluation sessions and the number of different foods accepted and tolerated at home. The clinician recorded the baseline number, and the post treatment and four-week follow-up numbers were recorded by his mother in a log and were reported to us via email.

Swallowing and eating outcomes were targeted via four different subcomponents: voluntary control of saliva swallows, rate of drinking, and aerophagia level. For evaluating voluntary control of saliva swallows, a latency measure was also utilized and consisted of the duration from a verbal command “swallow” to the time the swallow was initiated, as indicated by visual elevation of the hyolaryngeal complex and/or palpation of the complex by his mother (when visual inspection was not adequate). Rate of drinking was measured by the time taken to complete a 180 mL (6-ounces) amount of thin liquid through a straw and was measured in mL/minute. One hundred and eighty mL was the amount of liquid that was ingested by AB in a single feeding at the time of the initiation of the telepractice program. Therefore, it provided a measure of functional competence for a mealtime feeding. Aerophagia level was determined by the average amount (in liters) of air and saliva suctioned off of his feeding tube per day at three different time points (at baseline, post treatment, and at follow-up).

Quality of life was evaluated via the completion of a self-reported questionnaire (EAT-10) (Belafsky et al., 2008) completed by the mother. This tool has been validated for adult patients with swallowing problems, however, the questions were deemed relevant for pediatric cases as well. The wording of the questions on the EAT-10 was minimally modified so that the questions were addressed to the family and not the patient. The content and scoring remained exactly the same. Table 1 summarizes all the variables, how they were measured and when they were assessed.

Table 1. Outcome Variables, Assessment Measurements, and Time of Assessment

Outcome Variable	Measurement	Time of Assessment
Behavioral Variables		
<i>Oral acceptance of eating objects</i>	From time of command to time of placement of object inside patient's mouth (seconds) – <i>latency measure</i>	Baseline; Post-Intervention
<i>Oral acceptance of flavors/foods/consistencies</i>	# of different flavors/foods accepted and tolerated (swallowed)/week – <i>frequency measure</i>	Baseline; Post-Intervention; 4-week follow-up
Eating/Swallowing Variables		
<i>Voluntary control of saliva swallows</i>	From time of command "Swallow" to time of swallow initiation (seconds) – <i>latency measure</i>	Baseline; Post-Intervention
<i>Rate of drinking</i>	Amount of thin liquid in mL consumed via straw/minute – <i>rate measure</i>	Baseline; Session 2 and discontinued
<i>Aerophagia level</i>	Average amount of air and saliva (in liters) suctioned off of his feeding tube/day – <i>volume measure (family report)</i>	Baseline; Post-Intervention; 4-week follow-up
Quality of Life Variable		
<i>Swallowing-related quality of life</i>	Modified EAT-10 questionnaire – <i>rating scale (family report)</i>	Baseline; Post-Intervention; 4-week follow-up

INTERVENTION

The eight-session intervention followed consistent functional practice guidelines targeting the outcome variables, with use of selective compensatory strategies and motor learning strategies to support motor learning (Sheppard, 2013). Prior to each session, an email was sent to AB's mother with a detailed schedule of the session, including planned activities and a list of materials and foods required for the session. Table 2 presents all intervention strategies utilized and provides definitions and examples.

TELEPRACTICE ADAPTATIONS

Due to the mode of delivery, several telepractice adaptations had to be implemented. These are outlined below.

PARENTAL INVOLVEMENT

Parental involvement was required to facilitate treatment methods both during the sessions (Figure 1) and for generalization of skills during the home program. By participating as the session mediator in all sessions, AB's mother was incidentally trained in the routines to prepare for and conduct practice sessions. These included grading task difficulty as AB progressed and utilizing hyolaryngeal complex palpation in order to detect swallow completion. In addition, her knowledge of AB's preferences and tolerances was critical in helping the clinician select relevant and salient reinforcement strategies and develop effective treatment routines.

Table 2. Therapy Strategies

Therapy Strategies	Definitions	Examples in AB's sessions
Motor Learning Strategies		
Blocked Practice Sequence	Practice sequence in which individuals rehearse the same skill repeatedly (Schmidt & Wrisberg, 2004)	AB was encouraged to practice one task at a time for multiple repetitions, then rest and practice again.
Reduced Response Effort (Practicing Part/Simple Tasks)	Reducing the difficulty of a task by eliminating challenging components or practicing simple components first (Park, Wilde, & Shea, 2004)	AB practiced inserting a simulated spoon in his mouth prior to spoon-feeding.
Extrinsic Feedback (Knowledge of Performance and Results) [immediate and high in frequency]	Information provided to the child during, and after a task regarding the performance and the results of the task (Schmidt & Lee, 2005)	Frequent verbal acknowledgment of adequate performance was provided to AB for most tasks.
Reinforcement	Positive or negative reinforcement to increase the occurrence of a target behavior (Magoon & Critchfield, 2008)	Positive: A sticker was given to AB after each successfully completed activity Negative: Termination of a difficult task (aversive stimulus removed) once AB completed a designated number of trials (behavior)
New strategy combining aforementioned strategies ("Racing Car Swallow")	Systematic approach to teach voluntary physiologic saliva swallows as an alternative for the aerophagia, air-injection swallow.	Initial Steps: a) Cough b) Lollipop in mouth for 3 sec (later just simulated lollipop) c) Remove lollipop and pause for 3 sec d) Swallow saliva Steps were decreased systematically over time to step d
Strengthening and Compensatory Strategies		
Straw drinking	Sucking from a straw in order to improve oral pressure and orobuccal coordination (Nilsson, Ekberg, Olsson, Kjellin, & Hindfelt, 1996).	This was a well-developed skill for AB, which was used to introduce more viscous boluses (not yet accepted) and continue improving orobuccal coordination.
Chin Tuck	Swallowing while tucking the chin to facilitate a mature propulsion swallow as the base of tongue moves backwards (Logemann, 1998)	The chin tuck technique was incorporated in straw-drinking, spoon-feeding, and voluntary saliva swallows with AB as an added safety precaution and a means for better assurance that he was using a propulsion swallow.

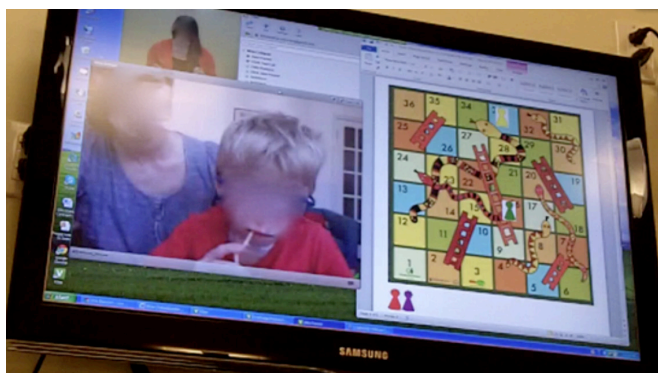


Figure 1. Telepractice therapy session. Image shows parent sitting next to child and facilitating session and board game shared via the VSee screen share utility. Clinician is shown in upper left hand corner.

PACING, INSTRUCTIONS AND REINFORCEMENT

To maintain the patient's attention and motivation level and to ensure a structured environment despite the distance, a consistent routine was used to introduce instructions and timing requirements for all activities. Instructions for the activities were presented via personalized PowerPoint slides, which were shared with the patient via the Screen Share utility of VSee. Timing requirements were targeted via a visual timer and computerized board games, which were controlled by the clinician (Figure 1). Via the Screen Share utility of VSee the patient could visualize all stimuli but had no direct control over them. He indirectly controlled them, however, with his performance (e.g., every successful attempt of a specific task resulted in the clinician moving a game piece one step further). Advancing the game and verbal confirmation provided feedback that acknowledged the successful completion of each task repetition. A sticker was used as feedback to acknowledge his successful completion of each practice activity. Although, AB appeared to find the treatment routines to be motivating in themselves, positive reinforcement was limited, at mother's request, to a surprise 'reward' (usually a small toy) that AB was given at the end of the session. All instructions and positive reinforcements were situational to AB, relating to his interests of favorite hobbies, musical artists, and games.

FOOD AND MATERIALS PREPARATION

All food and materials preparation was the responsibility of the family and the patient. This necessitated e-mail communication with the family one day prior to each session, which included specific instructions for the preparation of all materials. It also enhanced the principle of salience, because all sessions were conducted at the natural home environment and included foods and materials already familiar to the patient.

TECHNOLOGICAL ADAPTATIONS

In addition to the aforementioned adaptations, a few technology-related adaptations were also implemented. One 10-minute trial session was scheduled prior to the initiation of the program to test Internet connectivity and image and audio quality. Furthermore, the family was instructed to have an external camera and external microphone available in case visual and audio adjustments or zooming was necessary. During feeding trials, the patient was instructed to sit across from his mother and have his body lateral to the computer, in order for the hyolaryngeal complex to be visible during swallows. At times, a tape was placed over his thyroid notch to enhance visualization.

POST-INTERVENTION EVALUATION

The post-intervention evaluation was completed at the end of the eight-session program and included the same routines as the baseline evaluation. It also included a short satisfaction questionnaire that the family was asked to complete in order to rate their experience with telepractice. This questionnaire was based on a previous published questionnaire (Sharma et al., 2013).

FOLLOW-UP INTERVIEW (4 WEEKS POST-INTERVENTION)

Four weeks post-intervention a follow-up online interview with the mother was arranged. At follow-up, the mother was asked to complete the modified EAT-10 questionnaire, to list all foods currently consumed by AB, and to answer the following three open-ended questions: (a) On average how many liters of air/saliva per day are you suctioning off your child's feeding tube NOW? (b) How do you feel about your child's feeding and swallowing NOW compared to when you first came to the Dysphagia Research Clinic this summer? (c) What part (if any) of the therapy your child received this summer had the most positive impact on the changes you saw in his feeding and eating?

INTERVENTION FIDELITY – COMPLIANCE

At the end of each session, instructions were given to the patient's mother for the home program. Practice was required on an everyday basis and for at least 30 minutes. Fidelity was measured with weekly emails from his mother indicating the tasks the patient completed at home in between the tele-sessions. Review of the emails at the end of each week revealed that AB, with the aid of his mother, completed 98% of home program activities.

INTER-OBSERVER RELIABILITY

In addition, all sessions were video-recorded, and the two latency outcome measures (i.e., latency for oral acceptance of eating-related objects and latency for voluntary saliva swallows) were re-measured from the video recordings of the baseline and post-intervention evaluations by another judge (a certified SLP). A Pearson r correlation coefficient was calculated (Wessa, 2012) between the two raters' measurements and was found to be $r=0.9993$, indicating high correlation of their scores ($p<0.001$).

RESULTS

BEHAVIORAL RESULTS

Results of the latency measure used to determine oral acceptance/tolerance of eating-related objects at baseline and post-intervention show a post-treatment decrease in latency of 3.58 seconds for spoon acceptance (69% decrease) and of 11.09 seconds (62% decrease) for acceptance of a lollipop (Figure 2). Given AB's initial hesitation to perform these tasks at baseline, these results were based on one trial per item. Table 3 reports the different foods (and consistencies) AB consumed at baseline, post-intervention and at follow-up per mother's report and clinician's observations. This information suggests that AB was accepting to consume 5.25 times more foods at the end of the intervention and 11.25 times more foods at follow-up compared to baseline. Furthermore, his advancement into accepting a variety of pureed foods is evident post-intervention and at follow-up.

Table 3. Acceptance of Foods and Consistencies (Different Foods Accepted at Three Time Points)

	Baseline	Post-Intervention	Follow-up
Foods [consistency]	Neocate flavored [thin] Fortijuce [thin] Smoothies [thin] Actimel Yogurt Drink flavored [thin]	Neocate (Unflavored) [thin] Sweet Potato [puree] Sweet Corn [puree] Carrots [puree] Peas [puree] Spinach [puree] Butternut Squash [puree] Lentils [puree] Cheese [puree] Apples [puree] Bananas [puree] Mangoes [puree] Pear [puree] Peaches [puree] Parsnips [puree] Broccoli [puree] V8 Vegetable Juice [thin] Yogurt (Plain) [puree] Coconut Smoothie [nectar-thick] Coconut Kefir Smoothie [nectar-thick] Red Lentil Dahl [puree]	Oats [puree] Ready Brek [puree] Dried Apricots [puree] Raisins [puree] Prunes [puree] Molasses [puree] Butter [puree] Blueberry [puree] Sardines [puree] Tuna [puree] Rice [puree] Baked Beans [puree] Avocado [puree] Chocolate [puree] Kale [puree] Red Peppers [puree] Pumpkin seeds [puree] Sunflower seeds [puree] Papaya [puree] Maple Syrup [nectar-thick] Fresh Juice [thin] Cocoa [thin] Dessicated Coconut [puree] Nutritional Yeast flakes [puree]
# of different foods	4	21	24
# of all foods accepted	4	25	49

SWALLOWING/EATING RESULTS

Substantial reduction was seen in the latency measure on the voluntary saliva swallowing variable post-treatment, which showed a decrease of ~89% (Figure 3), and in aerophagia levels as measured by the average amount of liters suctioned off AB's feeding tube post-treatment and at follow-up (Table 4). Rate of drinking did not change through the four-week program and remained at ~32mL/minute. During session 2, AB demonstrated a sharp increase in rate to 180 mL/minute and thus this goal was discontinued as his baseline value was considered to be a preference and not a delay.

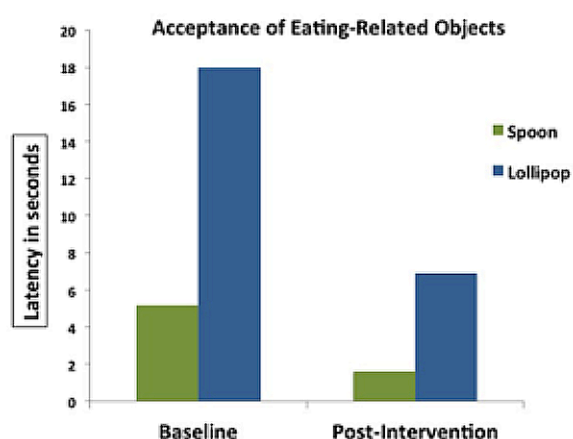


Figure 2. Latency of oral acceptance/tolerance of eating-related objects at baseline and post-intervention; results were based on one trial at each time.

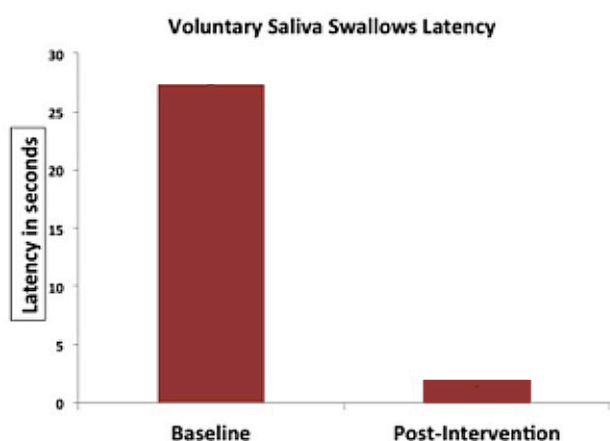


Figure 3. Latency of voluntary saliva swallows at baseline and post-intervention; results were based on one trial at baseline and on three trials post-intervention.

Table 4. Aerophagia Levels (Average Liters/Day Suctioned Off AB's Feeding Tube)

	Baseline	Post-Intervention	Follow-up
Total Amount/day	12 liters/day	<1 liter/day	<1 liter/day
# times/day	12	5	5

QUALITY OF LIFE AND TELE-SATISFACTION RESULTS

Quality of life was assessed at three time points (baseline, post-intervention, follow-up) (Table 5). The EAT-10 total score was improved by four points at post-intervention and by nine points at follow-up, indicating continuing improvements in quality of life even after the completion of treatment. More pronounced improvements were seen in relation to the item on globus sensation, which was perceived by the mother to be a "severe problem" (Score: 4) at baseline and post-intervention and was reduced to "no problem" (Score: 0) at follow-up. Mild improvements (1-2 point reductions) were also observed in the following parameters: going out for meals, pleasure of eating affected by the swallowing problem and swallowing is stressful. Swallowing solids and pills remained challenging. Results of the tele-satisfaction questionnaire are reported in Table 6 and show that the family appeared very satisfied by this program and mode of delivery.

Table 5. Responses to Modified EAT-10 (Belafsky et al., 2008) at Three Time Points

EAT-10 Question (summarized)	Baseline Score	Post-Intervention Score	Follow-up Score
1. Weight loss or gain	0	0	0
2. Going out for meals	4	2	2
3. Extra effort for liquids	1	1	1
4. Extra effort for solids	4	4	4
5. Extra effort for pills	4	4	4
6. Pain on swallow	0	0	0
7. Pleasure of eating affected	4	2	2
8. Globus sensation	4	4	0
9. Coughing during meals	0	0	0
10. Stressful swallowing	2	2	1
TOTAL Score	23	19	14

Note. Likert scale 0-4; 0=No Problem and 4=Severe Problem

Table 6. Modified Tele-satisfaction Questionnaire (Sharma et al., 2013) and Responses

Questions	Post-Intervention Responses
1. I was comfortable to have my child undergo an assessment for his swallowing/feeding disorder via the internet.	Strongly Agree
2. I was comfortable to have my child undergo rehabilitation for his swallowing/feeding disorder via the internet.	Strongly Agree
3. I had no difficulty seeing the online speech pathologist.	Strongly Agree
4. I had no difficulty hearing the online speech pathologist.	Strongly Agree
5. I would rate the online assessment and treatment as being equal to assessments and treatments conducted traditionally in the face-to-face method.	Strongly Agree
6. The instructions given during the online program were clear and easy to follow.	Strongly Agree
7. Telehealth can replace a face-to-face assessment and treatment of feeding and swallowing difficulties.	Agree
8. I would prefer to have a traditional (face-to-face) consultation with the speech pathologist despite possible costs and inconveniences.	Disagree

Note. Possible responses = Strongly Disagree, Disagree, Unsure, Agree, and Strongly Agree

DISCUSSION

The present case study reports the feasibility of an intensive closed-ended dysphagia telepractice program and its preliminary results in a pediatric patient with complex medical history and dysphagia. Our results suggest that this program was feasible.

Thus far, telepractice research in the area of dysphagia has focused on the adult populations (Georges, Belz, & Potter, 2006; Malandraki, McCullough, He, McWeeny, & Perlman, 2011; Malandraki et al., 2013; Perlman & Witthawaskul, 2002; Sharma, Ward, Burns, Theodoros, & Russell, 2013; Ward et al., 2009; Ward, Sharma, Burns, Theodoros, & Russell, 2012), and has reported promising results. To our knowledge this is the first study reporting the feasibility of telepractice in the rehabilitation of pediatric dysphagia. Few studies have examined the feasibility and validity of telepractice for speech and language in children, such as articulation disorders (Grogan-Johnson et al. 2011), stuttering (Lewis et al. 2008), and language assessment (Waite, Theodoros, Russell, & Cahill, 2008). With the rapid developments in technology and Internet connectivity, telepractice is becoming a popular endeavor for many clinicians, however studies examining feasibility, validity, reliability and treatment effectiveness are needed before certain practices are established. The present case study suggests that the use of telepractice in pediatric dysphagia treatment is feasible. Additionally, our preliminary results suggest that the treatment program designed for this specific patient was clinically effective.

Specifically, the treatment program was effective in improving five of six variables targeted during treatment. Furthermore, the results of the four-week post treatment follow-up interview with the patient's mother suggest

that many improvements seen in post treatment measures were retained. In addition, in several variables improvements were seen one month post-intervention. This suggests an additional carryover effect that may have been associated with on-going home practice or with generalization of behaviors through daily use.

Although, this is a single case study and our results need to be interpreted with caution, the rapid and substantial improvements seen in many of our variables and their maintenance four weeks post treatment are encouraging. We attribute these results to a number of factors, all of which were enhanced due to the use of telepractice.

First, the program used many principles of activity-dependent plasticity (Kleim & Jones, 2008), including high intensity and frequency of treatment with two hour-long therapy sessions per week and daily (at least) 30-minute practice at home for the duration of the program; saliency, with swallowing therapy occurring in his natural eating environment (home) with foods and liquids that the patient and his family prepared; and specificity, with multiple activities including swallowing and eating of a variety of foods and flavors as a method to improve swallowing skills. Evidence-based motor learning strategies (Schmidt & Lee, 2005), i.e., blocked practice, immediate feedback, positive and negative reinforcement, etc., were also employed in a very systematic manner for practicing all motor tasks targeted.

We believe that telepractice significantly enhanced these principles and strategies. Due to the distance, parental involvement was necessary and crucial for the sessions and for home practice. Online parent training occurred through explicit instruction prior to and following sessions and incidental instruction during her participation in the sessions. Pediatric dysphagia treatment typically focuses on the feeding relationship between the child and the parent (Davies et al., 2006). Although parental involvement has been shown to be important in treatment results of feeding and communication disorders (Buschmann, Jooss, Feldhusen, Pietz, & Phillippi, 2009; Davies et al., 2006; Millard, Nicholas, & Cook, 2008; Roberts & Kaiser, 2011), frequently in traditional face-to-face sessions parents may not have the time or drive to participate despite clinician's recommendations. When using telepractice for pediatric patients, the parents/caregivers are required to be mediators and facilitators of the sessions and thus they get directly involved and trained. Parental involvement and training during the sessions of AB enabled his mother to maintain the frequency and intensity of the program and encouraged everyday practice.

In addition, the parents were involved in all food selection and preparation including foods typically consumed at their home, increasing the saliency of this intervention. Another factor that enhanced saliency was the fact that this feeding program was conducted at AB's natural eating environment (home), an added

feature of task specificity, which may be another reason the effects of treatment were retained or continued to improve a month post treatment. Systematic written instructions, following a treatment structure routine, and specific reward systems had to be used to ensure the child's attention level and engagement in the therapy activities remained constant despite the distance. In typical in-person therapy sessions, instructions are more often verbal and may be different from day to day. This systematic and highly structured approach of therapy may have been further effective for AB given his diagnosis of autism (Schopler, Brehm, Kinsbourne, & Reichler, 1971).

Although most parameters showed improvements post intervention and at follow-up, a few remained unchanged or were discontinued. First, the family responses on the modified EAT-10 revealed unchanged swallowing difficulties with solids and pills. Although gains had been made in intake of small amounts of puree foods, it appears from this result that the family did not see this as notable in terms of the EAT-10 question. In part this may reflect semantic differences in what constitutes a 'solid' food.

Rate of drinking for liquids was a parameter that was discontinued after the second session. This variable was targeted for the first two sessions to decrease time and effort of eating, given that AB's rate at baseline was considered to be moderately slow. This was considered to be a reflection of limitation in swallowing efficiency. During session 2, AB demonstrated a sharp increase in rate to 180 mL/minute (from 32mL/minute). At that time, exercises targeting bolus size for liquid swallows by straw were discontinued based on the judgment that AB had achieved capability for larger and more efficient liquid bolus swallows. The baseline slower mealtime rate could, therefore, be considered a preference rather than limitation. The treatment focus was shifted to targeting skills related to spoon-feeding of puree consistencies.

Despite these few parameters, overall, AB's eating and feeding skills and quality of life were improved and continued to show progress at follow-up. Additionally, one of his severe symptoms, causing significant discomfort to him and the family, was aerophagia. Aerophagia levels dropped drastically post treatment, and this is attributed (by both the clinicians and the mother) to his newly developed ability to successfully perform voluntary saliva swallows. At baseline AB appeared very apprehensive of attempting to swallow his saliva upon command and was observed to frequently exhibit an audible air swallow. The systematic approach we used to teach him to stimulate saliva production and then voluntarily swallow his saliva enabled him to develop a more mature swallow pattern and reduce the frequency of air swallows. According to the parents and their responses to the open-ended questions during the four-week follow-up interview, the practice of the voluntary swallow of saliva was the part of treatment that had the most impact on AB's swallowing and feeding progress.

In addition, AB's parents appeared rather satisfied with the use of telepractice and rated the online rehabilitation approach as being equal to the traditional in-person approaches. They further stated that they preferred this method of delivery for their son's feeding services compared to an in-person method. These perceptions are in agreement with results of several previous investigations reporting high levels of satisfaction especially at the post-assessment or post-treatment phase, when patients have also experienced the benefits of treatment (Cranen, Veld, Ijzerman, & Vollenbroek-Hutten, 2011; Demiris, Speedie, & Finkelstein, 2001; Sharma, et al. 2013). According to Sharma and colleagues, this also may be associated to increased confidence with technology achieved post participation in a telepractice program (Sharma et al., 2013).

LIMITATIONS

Despite the promising results of the present case report, there are limitations that need to be considered. First, this is a single case, which significantly limits the generalizability of the results and of this telepractice program. A study with larger sample size testing two modes of treatment (telepractice vs. in-person) would be necessary before these findings could be generalized to a telerehabilitation protocol for pediatric dysphagia intervention. Furthermore, many of our variables measurements were based on a small number of trials of a specific task and thus, valid pre-post statistical comparisons were not possible. The main reason for this was AB's difficulty or severe hesitation to complete a task (especially during the baseline evaluation), and our desire to complete the assessments without frustration or early dismissal or the early feeling of satiety, which would not allow for additional food trials to be tested.

Furthermore, some of the measurements we completed were based on the mother's responses and thus may include bias. We had no way of testing the reliability and validity of her responses, other than with our own observations of AB's behavior during the treatment sessions and his improving performance. Additionally, the mother was asked to complete online questionnaires, and this request was made by a research assistant (RA) who was not involved in the evaluation or treatment of the child. This RA was in direct contact with the mother via e-mail and the RA transferred the data to an excel form. Despite this process, some bias in the mother's responses is probably expected, since it was not explicitly explained to her that the clinicians would not have access to her responses. In the future, in addition to having a blinded RA collecting these data, the parents and patients should also be informed that no information would be transmitted to the clinicians.

The use of the modified EAT-10 questionnaire was solely decided based on the fact that it is a simple and efficient

swallowing-related quality of life questionnaire, and such tools are not yet available for the pediatric population. Validation of the adapted version and standardization is important for the use of this tool in future studies.

Additionally, the complex medical history of AB and the inclusion of several treatment strategies in our protocol complicate the interpretation of the findings. Specifically, it is difficult to determine which treatment technique was effective for each developed or emerging skill. However, our goal was to provide AB with optimal care using an online treatment delivery mode, and not to test the effectiveness of specific treatment strategies. Future research is needed to provide more evidence on specific treatment techniques used in our study. Last, technical challenges including video quality compromise were evident infrequently (during one session), but were easily alleviated with technical support.

CONCLUSION

The present case report shows the feasibility and preliminary effectiveness of an intensive telepractice program in the treatment of pediatric dysphagia. To our knowledge this is the first study reporting the feasibility of telepractice in the rehabilitation of pediatric dysphagia. Given the large number of pediatric and adult patients with swallowing and feeding disorders around the globe, the variable expertise available and the existence of underserved populations, the need for establishing evidence-based telepractice programs for dysphagia assessment and treatment is high. Such programs need to be developed with the appropriate expertise and careful consideration of patient and family needs, clinician's qualifications, technological requirements and technical support. The preliminary results of the present case study show promise for both pediatric assessment and treatment, however, further investigation is warranted to determine whether this mode of delivery can be generalized and is valid and reliable.

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